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Cor pulmonale due to untreated asthma in a young woman with coexistent anorexia nervosa

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Introduction

Asthma rarely presents as cor pulmonale (1,2), although the presence of hypoxaemia, hypercapnia, fluid retention and ECG evidence of right heart strain has been documented (3). In previous reports, the diagnosis of cor pulmonale has initially caused difficulty with the underlying disease unknown to the patient or the attending physician. We report a case of cor pulmonale in a young woman in whom reversible airway disease was suspected, but initially she failed to comply with investigation or treatment.

Case Report

A 22-year-old female maths graduate who did not smoke, presented to the outpatient clinic having been diagnosed with end-stage respiratory failure, requiring continuous oxygen therapy. In addition to her respiratory disease, she suffered from the eating disorder, anorexia nervosa, and initially refused any investigation or treatment for either condition.

As a child, she had experienced episodes of wheezing after exertion and upper respiratory tract infections. There had been loss of school attendance due to respiratory problems, however she obtained 9 grade A's at 'O' level and 3 'A' levels leading to a first class honours degree in Mathematics. Her father, who suffered from asthma and whose fingers were clubbed, had taken her to his physician in the U.S.A. where he lived.

In the U.S.A., investigations revealed a forced vital capacity (FVC) of 1·28 l, a forced expiratory volume in 1 s (FEV_1) of 0·51, a ratio of FEV_1 to FVC (FEV_1%) of 39 and arterial blood gases; pH 7·38, pO_2 of 42 mmHg, pCO_2 of 64 mmHg, 71% O_2 saturation. Two sweat chloride tests were within normal limits. She had mild eosinophilia, normal white and red cell count. Immunoglobulins showed a raised IgG and IgA. Aspergillus precipitins were negative.

A diagnosis of end-stage respiratory disease was made and she was prescribed oral theophylline mixed with peppermint cream and long-term oxygen, the only treatments to which she would agree. Initially, she had been referred to an eating disorder clinic as she was underweight and had secondary amenorrhea for 2 yr (her menarche occurred aged 14 years).

On examination, she was 5′ 1″ tall, weighed 5 stone and wore her hair in two long plaits that reached her thighs. She was cyanosed, clubbed and had lanugo. Her pulse was 110 beats min^{-1}, blood pressure was 110/70 mmHg, jugular venous pressure was not elevated, there was a right ventricular heave and a loud pulmonary component to the second heart sound. Her chest was over-inflated, she was using her accessory muscles of respiration, the respiratory rate was 30 breaths min^{-1} and auscultation revealed bilateral expiratory wheezes and right lower lobe crackles. The liver was not enlarged nor was any peripheral oedema present. Initial investigations showed FVC of 0·95 l (29%), FEV_1 of 0·39 l (13%) and FEV_1% of 41. The chest radiograph revealed over-distension of the lung fields and elevation of both hilar shadows. Her electrocardiogram (ECG) showed sinus rhythm, 'P' pulmonale, right axis deviation and right ventricular hypertrophy with strain (Fig. 1).

A diagnosis of cardio-respiratory failure secondary to asthma was made and admission advised to assess the degree of reversibility and to institute treatment. She declined admission and in view of her anorexia nervosa, a psychiatric opinion was advised. The patient and her mother were against any involuntary admission for either condition on the grounds that this would represent a loss of the patient's liberty. One month later, she voluntarily entered the ward when her examination was unchanged. Her spirometry showed FVC of 0·54 l (16·3%), FEV_1 of 0·23 l (7·9%) and FEV_1% of 43. After 5 mg nebulized salbutamol, these rose to 1·00 l, 0·45 l and 45 (increases of 85%, 95% and 2% respectively). Her

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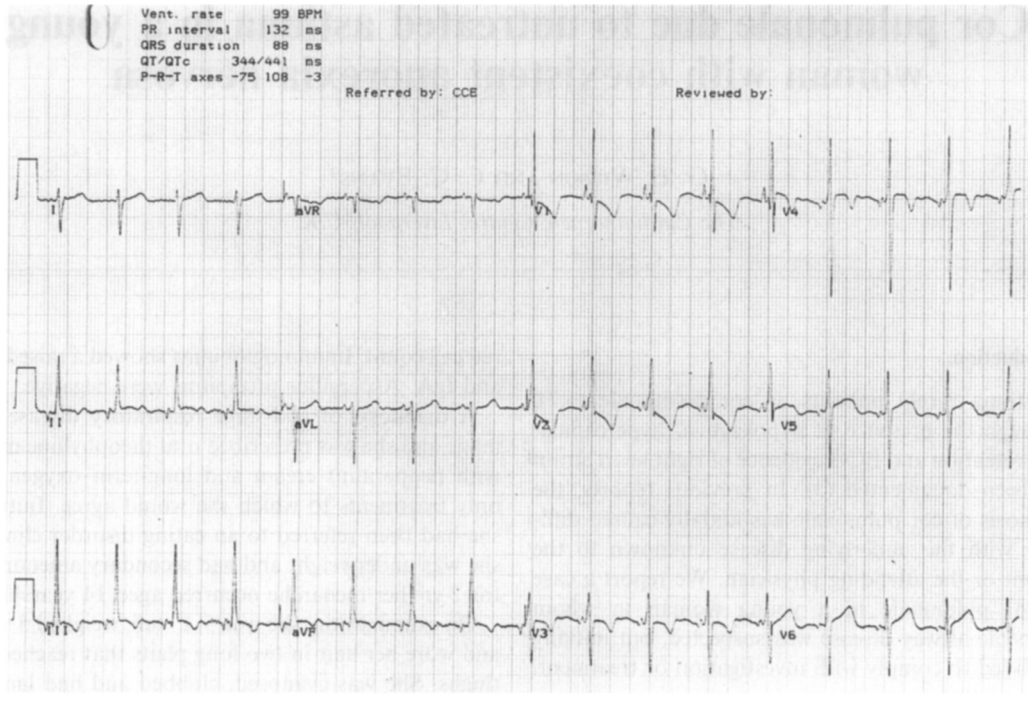

Fig. 1 ECG showing 'P' pulmonale, right axis deviation and right ventricular strain.

arterial blood gases on 2 l of oxygen showed pH 7·44, pO_2 of 18·8 kPa and pCO_2 of 7·3 kPa. Her peak flow at the time of admission was 30 l min^{-1} pre-nebulized salbutamol (5 mg) and 50 l min^{-1} post-nebulizer. Echocardiography showed a dilated right ventricle and mild tricuspid regurgitation. Her skin prick tests were negative to common allergens. Other investigations revealed: haemoglobin 11·1 g dl^{-1}, white cell count 4·5 × 10 (neutrophils 59%, lymphocytes 6%, monocytes 27%, eosinophils 8%), ESR 85 mm in the first hour. Sodium, potassium, urea, creatinine, albumin, calcium, bilirubin, alkaline phosphatase, aminotransferase and lactate dehydrogenase were all normal. Total protein, globulin and phosphate were all mildly raised. Protein electrophoresis showed a marked increase in alpha 1 globulin and a moderate increase in gamma globulin. Aspergillus precipitins were negative.

Her treatment consisted of 40 mg oral prednisolone once a day, 5 mg nebulized salbutamol once every 4 h and 0·5 mg of nebulized iprotropium bromide once every 6 h. Her overall peak flow improved on this regime although a diurnal variation was noted. Her afternoon values pre- and post-nebulizer were 270 l min^{-1} and 300 l min^{-1} respectively, morning values were 180 l min^{-1} and 220 l min^{-1} respectively. Additional inhaled treatment was started with salmeterol 50 μg twice daily and beclomethasone dipropionate 1000 μg twice daily (through a volumatic device).

A repeat of the arterial blood gases, after treatment, in air showed pH 7·465, pCO_2 of 6·95 kPa and pO_2 of 12·05 kPa.

In view of her improved blood gases, oxygen therapy was discontinued and she was discharged home and reviewed in clinic. Two years later, her asthma is well controlled on prednisolone 5 mg once a day, salbutamol 400 μg four times daily, salmeterol 100 μg twice daily, and beclomethasone diproprionate 800 μg twice daily. Her spirometry is FVC of 2·27 l (69%) and FEV_1 of 1·62 l (56%) with daily peak flows of between 200–300 l min^{-1}. She currently attends university and goes canoeing, trampolining, skiing and plays the flute. She writes computer programmes including one to monitor her peak flow. She has attended the eating disorder clinic, her weight is now 7 stone 3 pounds and her menstrual cycle is normal. Her ECG has shown resolution of some of the previous abnormalities (right axis deviation, large R waves in V1–3) although her echocardiogram shows some right ventricular enlargement.

Discussion

The presentation of asthma as cor pulmonale has previously been reported, as has the response to

treatment, including a reversal of ECG changes. In this case, it was the reluctance of the patient and her mother to further investigation and treatment, which caused the initial delay in diagnosis and treatment.

Anorexia is a potentially fatal illness with a 20% mortality at 20 yr follow-up (4). The recovery in this patient's cardio-respiratory failure has been accompanied by an improvement in her nutritional status. The explanation for this is complex. Admitting her to hospital may have reduced vomiting, separated her from any laxatives, and corticosteroids would have altered her appetite regulation. Moreover, her improved nutritional status may have resulted from her improved cardio-respiratory function as cachexia may result from advanced cardiac or respiratory failure (5).

A relationship is also said to exist between chemoreceptor sensitivity and extroversion in healthy young women (6), and also it has been suggested that mixed venous pCO_2 is higher in introverts than extroverts (7).

Whatever the interplay between anorexia nervosa and the underlying cor pulmonale, asthma remains an important treatable cause of the latter.

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